



Prevalence of Malocclusion Patterns in Mouth Breathing Children Compared to Nasal Breathing Children – A Systematic Review

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Abstract: The prevalence of malocclusion in mouth breathing children compared to nasal breathing children is unclear, therefore the aim of this study is to identify this prevalence in a systematic review. Seven studies were included. Six studies investigated the prevalence of malocclusion in mouth breathers and compared them with the prevalence data in nose breathers. One study evaluated the prevalence of mouth- and nose breathing in children with malocclusion. The prevalence rates for class I to class III malocclusions vary significantly between studies. For class I malocclusions, a prevalence between 9% and 74% was identified in the included studies. For class II malocclusions a prevalence between 21% and 73% was shown, which equalled roughly the prevalence of class I malocclusions. Class III malocclusions were the least prevalent, between 5% and 24%. There is insufficient evidence for a difference in prevalence patterns of class I – III malocclusions between mouth and nasal breathers in order to draw a definite conclusion. The quality of the included studies was rated moderate to poor. Research on the prevalence of malocclusions in mouth breathing compared to nasal breathing children is scarce. Identified prevalence rates differ significantly, which is likely due to different assessment and reporting methods used in the included studies. The evidence of a difference in prevalence patterns for class I – III malocclusions and other occlusion traits between mouth and nasal breathers is insufficient to draw a definite conclusion.

Keywords: Occlusion, Mouthbreathing, Nosebreathing, Prevalence, Malocclusion

1. Introduction

Malocclusion is described as the abnormal alignment of the teeth, which considers the relationship of the upper and lower teeth being fitted together [2]. In normal occlusion, the upper teeth slightly overlap the lower teeth, which allows the teeth to fit into the corresponding fossae of the opposite teeth [2]. Malocclusion is common but is not considered an unnormal or unhealthy condition [3]. It may be aesthetically unfavorable, but it is not to be seen as a need for treatment per se, rather it should be assessed according to the dental health status and need for correction, i.e. through assessment of the abilities to chew, talk, breathe and swallow [1]. The WHO doesn't define malocclusion as a disease but rather a set of dental deviations which potentially can influence the quality of life [3].

Mouth breathing or the process of inhaling and exhaling through the mouth instead of the standard nasal breathing is estimated to affect between 25% and 57% of children [5-7]. Excessive mouth breathing is characterized by the insufficient warming of the air compared to nasal inhalation, which may lead to dryness of the mouth due to bypassing the nasal canal and the paranasal sinuses [8]. In patients with mouth breathing, strong associations have been found with a number of oral and medical conditions, such as halitosis, obstruction, and other abnormalities of the upper respiratory airways as well as congestions [9-12].

1.1. Relationship Between Mouth Breathing and Malocclusion

During growth the craniofacial structures adapt to the altered breathing patterns in mouth breathers, leading to

changes in the facial musculature affecting the dental arches and positions of the teeth, structural dispositions of the tongue, lips, palate, and mandible as well as face deformity [11]. Likewise, the chewing activity in mouth breathers can be decreased, negatively affecting the vertical position of the posterior teeth, which can lead to malocclusion [20, 21].

Facial deformities have also been reported in connection with mouth breathing, being characterized by long faces with an increase in the anterior lower facial height, increased palate height, tooth crowding, a narrow nasal passage and enlarged nostrils [17, 18, 22, 23].

A recent systematic review and meta-analysis on the effects of mouth breathing on facial development and malocclusion suggests that mouth breathing can cause underdevelopment of the mandible, with backwards and downward rotation of the mandible and steep occlusal plane. Additionally, a tendency of lip inclination of the anterior teeth, as well as airway stenosis, was found in mouth breathers [16].

The study of the relationship between mouth breathing and the frequency of malocclusion has produced conflicting results, with Leech et al. (1958) suggesting that mouth breathing not having any influence on dentofacial morphology, being mirrored by the studies of Gwynne-Evens and Ballard (1959) which observed that mouth breathers did not present with changes in jaw growth, malocclusion and other anomalies of dental position [24, 25]. Other studies found a relationship between mouth breathing and an increased frequency of malocclusion patterns. For example, a systematic review investigating whether mouth breathing children are more likely to present with malocclusion compared to nasal breathing children has shown that the prevalence of Angle Class II malocclusion was higher than Class I malocclusion in mouth breathing children [26]. For this review, however, most included studies were published in Portuguese, and the few English language studies were published a decade ago.

1.2. Aim of this Review

Given the controversy regarding the relationship between malocclusion and mouth breathing, this systematic review aims to update and extend on previous findings on the relationship between mouth breathing and the frequency of malocclusions compared to nasal breathing. The research question for this review is:

What is the prevalence of malocclusion patterns in mouth breathing children compared to nasal breathing children?

Does the prevalence differ between mouth and nasal breathing children?

2. Methods

2.1. Study Design and Database Search

This study was a systematic literature review, with a literature search being conducted from Database start up to the 15th July 2020, using the following databases, search interfaces, and other search methods: MEDLINE (via Pubmed), Cochrane Library, Google Scholar, bibliographic searches of the included studies.

Inclusion criteria were studies which included healthy children aged 0-18 years with mouth breathing compared to nasal breathing children and measured prevalence rates of malocclusion OR children with a diagnosis of malocclusion and assessment of outcome measures of mouth and nasal breathing, studies published in peer-review journals in English; and observational study designs, such as case-control studies, cross-sectional studies or cohort studies. The choice of the study design is justified as development of malocclusion in conjunction with mouth breathing will not be able to be assessed in randomized clinical trials.

Exclusion criteria were children with systemic disease, oral or maxillofacial trauma, expert opinion articles, reviews, letters, editorials, and conference abstracts.

A summary of the eligibility criteria is given in Table 1.

Table 1. Eligibility criteria for study inclusion.

Criteria	Inclusion	Exclusion
Study design	Observational studies (case-control studies, cross-sectional studies or cohort studies)	Expert opinion articles, reviews, letters, editorials, and conference abstracts
Population	Studies that assessed healthy children aged 0-18 years; diagnosis of mouth/nasal breathing OR diagnosis of malocclusion	Children with systemic disease, oral or maxillofacial trauma
Outcome	Prevalence rates of malocclusion in mouth breathers vs nasal breathers OR Prevalence rates of mouth breathers vs nasal breathers in children with malocclusion	Other outcomes
Study language	English	Other languages

2.2. Study Selection and Data Extraction

The author of this study initially selected the articles identified through the database search based on the predefined inclusion criteria and titles and abstracts. Full-text articles of potentially eligible studies were obtained evaluated for final inclusion in the review, based on the eligibility criteria specified in Table 1. The data extraction of the included studies was performed into several

predefined data extraction tables. The following information was obtained: study characteristics (Author, year, study country, number of subjects, study design, age range of subjects, characteristics of study participants), methods of diagnosis of malocclusion and mouth breathing, and the prevalence rates of malocclusion in MB vs NB, or the rates of MB vs NB in children diagnosed with malocclusion.

2.3. Quality Assessment of the Included Studies

The quality assessment of the included studies was conducted with the critical appraisal tools for cross-sectional and case-control studies of the Joanna Briggs Institute (JBI) [27]. These assessment tools have been developed by the JBI and collaborators and approved by the JBI Scientific Committee and have been recommended as a suitable assessment method in a variety of study designs [28]. The quality is evaluated in four domains: participant recruitment, exposure, confounding, and outcomes [27].

2.4. Reporting of this Review

The reporting of this present systematic review is based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [29].

3. Results

A total of 826 studies were identified, of which 795 studies were excluded based on title and abstract. After the evaluation in full text total of seven studies, including a total of 2,554 patients, were included in this review (Figure 1).

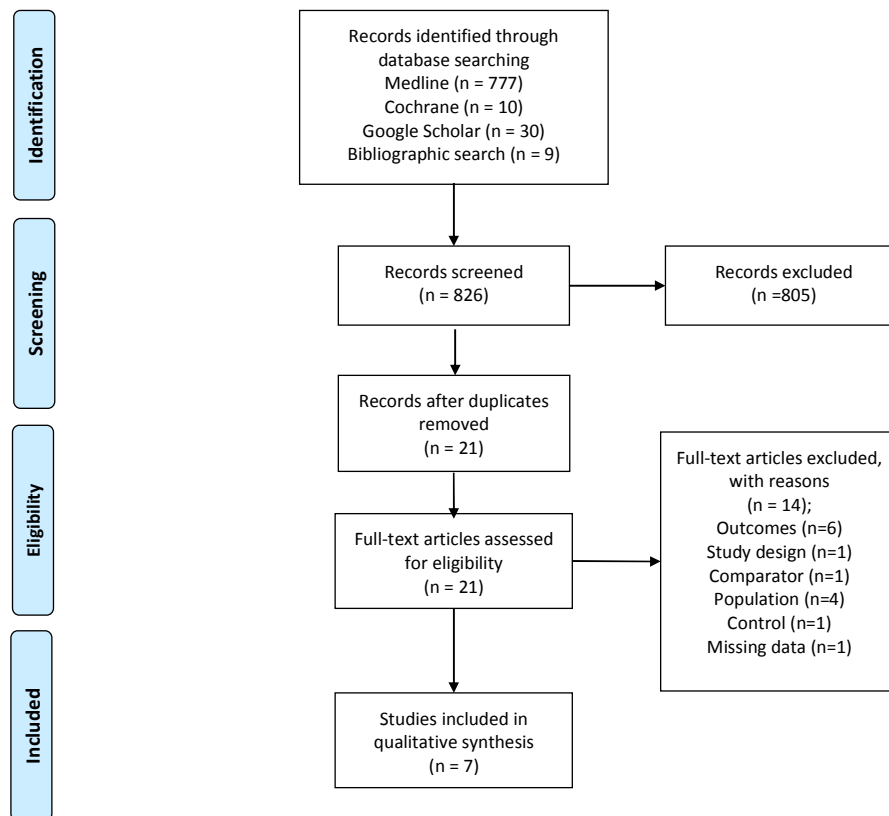


Figure 1. PRISMA Flowchart.

3.1. Summary of findings

The prevalence rates for class I to class III malocclusions vary significantly between studies. For class I malocclusions, a prevalence between 9% and 74% was identified in the included studies. For class II malocclusions, a prevalence between 21% and 73% was shown, which equalled roughly the prevalence of class I malocclusions. Class III malocclusions were least prevalent, between 5% and 24% were shown (Table 2).

Results show that in mouth breathers one out of five studies assessing the prevalence of class I – III malocclusion showed a significant difference for class I-III malocclusions compared to nasal breathers (Zicari 2009) [30]. This study had a moderate to poor quality rating. Another study (Rossi 2015) [13] showed a statistically significant difference between mouth and nasal breathers only in the prevalence of class II malocclusions, which also showed a moderate to poor

quality rating. Two studies did not show a statistically significant difference between mouth and nasal breathers for the occurrence of class I-III malocclusions. One further study showed trends of differences in malocclusion patterns between mouth and nasal breathers but did not conduct a statistical analysis (Table 3).

In children with malocclusions, only one study reported results on a significantly higher prevalence of mouth breathing compared to nasal breathing but did not differentiate between different classes of malocclusion.

Hence, there is insufficient evidence for a difference in prevalence patterns of class I – III malocclusions between mouth and nasal breathers in order to draw a definite conclusion. Additionally, there was also insufficient evidence for other occlusion traits in mouth breathers compared to nasal breathers to make a firm conclusion (Table 4).

The moderate to poor quality rating of the included studies

can be attributed to insufficient reporting of eligibility criteria and participant recruitment as well as consideration for confounding variables.

Table 2. Prevalence of malocclusion*.

Malocclusion Type	Class I	Class II	Class III
Included studies	9% to 74%	21% to 73%	5% to 25%

* Based on five studies: D'Ascanio (2010) [32], Gois (2008) [4], Harari (2010) [31], Pacheco (2015) [14], Rossi (2015) [13], Shanker (2004) [33], Zicari (2009) [30]

Table 4. Summary of findings for other types of malocclusions, MB vs NB.

Malocclusion Type	Overbite	Crossbite	Open bite	Deep bite	Overjet
MB vs NB	-	++	(+)	(+)	-

++ significant difference to nasal breathers in most of the studies, (+) significant difference to nasal breathers in half of the studies; - no significant difference to nasal breathers in one study

MB: mouth breathers; NB – nasal breathers

3.2. Study Characteristics

The included studies comprised six case control studies and one cross-sectional study. The total number of participants in the included studies was 2554 patients, of which 541 children were classified as mouth breathers, and 750 were nasal breathers. The countries of study conduct were Brazil, Italy, USA and Israel. The age of the included children ranged from 3 to 18 years. One study (Shanker 2004) [33] was a longitudinal study with a follow-up time-period of 4 years, which included four yearly assessments. One study (Gois 2008) [4] included children with malocclusion and investigated the rates of nasal vs. mouth

Table 3. Summary of findings for class I-III malocclusions, MB vs NB*.

Malocclusion Type	Class I	Class II	Class III
MB vs NB	--	(+)	--

(+) significant difference to nasal breathers in half of the studies; -- no significant difference to nasal breathers in most studies

MB: mouth breathers; NB – nasal breathers

* Based on five studies: D'Ascanio (2010) [32], Gois (2008) [4], Harari (2010) [31], Pacheco (2015) [14], Rossi (2015) [13], Shanker (2004) [33], Zicari (2009) [30]

breathing compared to children without malocclusion, whereas in the remaining studies, the rates of malocclusion were assessed in the recruited mouth breathers compared to nasal breathers (Table 5).

3.3. Methods of assessment

The methods of assessing malocclusion were not reported in one study (Pacheco 2015) [14] and were, in general, poorly described and were different for each study. The same trends were observed for the assessment of mouth breathing, where each included study used a different method, and they were generally poorly reported. (Table 6).

Table 5. Study characteristics.

Study	Study Design	Study Country	N	Age	Subjects
D'Ascanio 2010 [32]	Case Control Study	Italy	196	Mean age 8.8 years; age range 7–12 years	Group 1: 98 children with obligate mouth-breathing secondary to nasal septum deviation Group 2: 98 age- and sex-matched nasal breathing controls
Gois 2008 [4]	Case Control Study	Brazil	300	Mean age 4.52 years; range 3-6 years	Group 1: 150 individuals with at least one of the following malocclusions: anterior open bite, posterior crossbite, or overjet of more than 3 mm. Group 2: 150 individuals without malocclusion
Harari 2010 [31]	Case Control Study	Israel	116	Mean age 12.5 years; range 10-14 years Age range 7-12 years old, with the majority aged between 8 and 9 years old (42.9%)	Group 1: 55 paediatric mouth breathing patients Group 2: 61 patients were normal nose breathers
Pacheco 2015 [14]	Cross-sectional Study	Brazil	687		Group 1: 167 children were mouth breathers Group 2: 520 children were nose breathers
Rossi 2015 [13]	Case Control Study	Brazil	966	Age range 5-18 years	Group 1/Group 2: Consecutive sample of mouth and nose breathing patients of both sexes Unclear how many were nose breathers and how many were mouth breathers
Shanker 2004 [33]	Case Control Study	USA	147	Mean age 8.95 years; range 8-12 years	Group 1/Group 2: Normal and healthy children who were not taking any medication and who were between 8 and 10 years old, who were followed for 4 years Unclear how many were nose breathers and how many were mouth breathers
Zicari 2009 [30]	Case Control Study	Italy	142	Age range 6-12 years	Group 1: A paediatric population of 71 mouth breathers Group 2: 71 age- and sex-matched nasal breathers without allergies

Table 6. Methods of diagnosing malocclusion and mouth breathing.

Study	N	Subjects	Method of assessing malocclusion	Method of assessing mouthbreathing
D'Ascanio 2010 [32]	196	Group 1: n=98 MB Group 2: n=98 NB	Anamnesis, clinical examination, and cephalometric analysis by standard lateral cephalometric X-ray	Not reported
Gois 2008 [4]	300	Group 1: n=150 with malocclusion	Clinical examination by a unique previously calibrated orthodontist: measurements of overjet and overbite, classification of primary	Assessment of lip incompetence, dry lips, and fogging on the lower side of a

Study	N	Subjects	Method of assessing malocclusion	Method of assessing mouthbreathing
		Group 2: n=150 without malocclusion	canine and second molar relationships, and presence or absence of anterior crossbite, posterior crossbite, and anterior open bite in centric occlusion using a tongue blade and a mechanical pencil to record the amount of overjet and overbite, using the Warren and Bishara criteria (Warren and Bishara 2002) [41] Criteria for Class I, Class II, or Class III malocclusion and for posterior and anterior crossbite were based on a method previously described by Foster and Hamilton (Foster and Hamilton 1969) [39]	double-faced mirror in agreement with Moyers' criteria (Moyers 1973) [40] and used by others (Bresolin 1984, Hartgerink 1989) [37, 38]
Harari 2010 [31]	116	Group 1: n=55 MB Group 2: n=61 NB	Comprehensive dental examination, a cephalometric analysis, and dental study models. The study models were analysed and measured for symmetry, arch form, tooth position, occlusion, and other normal parameters liable to undergo changes due to mouth breathing; cephalometric X-rays	Case history and complete physical examination, including anterior rhinoscopy, flexible nasopharyngoscopy, or lateral nasopharyngeal x-ray, and confirmed by questionnaire answered by the parents The mirror test was performed by placing a graded mirror under the nose, and a halo of water vapor was marked after the third normal expiration. Two lip Seal tests were also performed for three minutes each, one using a sticky tape to close the lips and the other with water into the child's mouth
Pacheco 2015 [14]	687	Group 1: n=167 MB Group 2: n=520 NB	Not reported	Through patients' predominant breathing pattern history and the findings on clinical examination based on Wieler (2007) [42] Modified Simultaneous Nasal and Oral Respirometric Technique, nasal resistance measurement, those showing >80% of nasality were categorized as nasal breathers, the rest was designated as mouth breathers (based on Method of Warren et al., 1990) [43]
Rossi 2015 [13]	966	Group 1/Group 2: MB and NB	Plaster model, cephalometric x-ray, based on Angle classification	Rhinomanometry in accordance with the International Standardization Committee on Objective Assessment of the Nasal Airways and standardized questionnaire
Shanker 2004 [33]	147	Group 1/Group 2: Normal and healthy children	Intraoral measurement by Boley Gauge Intraoral examination (dental class type, overbite, overjet, midlines, crossbite, and presence of parafunctional oral habits such as atypical swallowing, labial incompetence, finger sucking and sucking of the inner lip); latero-lateral projection teleradiography; orthopantomogram; alginate impressions of the dental arches; evaluation of the study casts and cephalometric analysis	
Zicari 2009 [30]	142	Group 1: n=71 MB Group 2: n=71 NB		

MB: Mouth breathers; NB: Nasal breathers

3.4. Prevalence of Malocclusion/Mouth Breathing/Nasal Breathing

Six studies reported on the prevalence of malocclusion in recruited mouth breathers vs nasal breathers, whereas one study (Gois 2008) [4] included children with malocclusion and investigated the rates of nasal vs. mouth breathing compared to children without malocclusion (Table 7).

For class I – III malocclusion, five studies reported prevalence results in mouth breathing and nasal breathing children. Of these, one study (D'Ascanio 2010) [32] failed to provide statistical test results for significance, although the differences were markedly seen, whereby in nasal breathers class I malocclusion (normal occlusion) was the most prevalent (79%), and in mouth breathers class II malocclusions was the most prevalent (66%), followed by class III malocclusion (24%). Two further studies did not show a statistically significant difference between mouth and nasal breathers for the occurrence of class I-III malocclusions (Harari 2010; Pacheco 2015) [14, 31]. One study showed a significant difference between mouth and nasal breathers for all class I-III malocclusions, whereby in nasal breathers class

I malocclusion was the most prevalent (79%) and in mouth breathers class I and II malocclusions occurred almost equally (52 vs. 44%) (Zicari 2009) [30]. Rossi et al. (2015) [13] showed a statistically significant difference between mouth and nasal breathers only in the prevalence of class II malocclusion (64% vs. 48%, respectively) (Table 7).

In the longitudinal study by Shanker et al. (2004) [33] with a follow-up of 4 years, there was no statistical difference seen between mouth and nasal breathers in the prevalence of overbite over the four follow-up time points (Table 7).

Other aspects of malocclusion showed also different results. In the study by Harari et al. (2010) [31], posterior crossbite was significantly associated with mouth breathing. Likewise, crossbite, open bite, and deep bite were also significantly associated with mouth breathing in the study by Zicari et al. (2009) [30]. The study by Pacheco et al. (2015) [14], showed a similar prevalence of deep overbite, anterior open bite, posterior crossbite, pronounced overjet, and atresic palate between mouth and nasal breathers (Table 7).

In patients with malocclusion, the prevalence of those displaying mouth breathing was higher than those with nose breathing, whereas in those patients without malocclusion,

nasal breathers were more prevalent, resulting in a ten times higher chance of mouth breathers to suffer from malocclusion compared to the nasal breathers (Gois et al., 2008) [4] (Table 7).

Table 7. Prevalence of malocclusion in MB vs NB or MB vs NB.

Study	Prevalence Malocclusion	Prevalence Mouthbreathing	Adjusted OR (CI, 95%)	Conclusions
D'Ascanio 2010 [32]	Normal occlusion: MB (9%) vs NB (79%) Class II occlusion: MB (66%) vs NB (17%) Class III occlusion: MB (24%) vs NB (4%)	N/A	NR	Most mouth breathing patients displayed a class II skeletal malocclusion, while most nasal breathing control subjects showed normal occlusion
Gois 2008 [4]	N/A	Patients with malocclusion: 30% mouth breathers and 20% nasal breathers Patients without malocclusion: 7% mouth breathers and 43% nasal breathers	MB vs NB: 10.9 (5.5–21.4), p=0.000	Mouth breathing children had 10.9 times greater chances of having malocclusion than children who breathe through the nose
Harari 2010 [31]	Class I occlusion: MB (22%) vs NB (31%) - ns Class II occlusion: MB (73%) vs NB (62%) - ns Class III occlusion: MB (5%) vs NB (7%) - ns Posterior crossbite: MB (49%) vs NB (26%) - p=0.006 Compared to the NB, the MB group presented a similar prevalence of deep overbite, anterior open bite, posterior crossbite, pronounced overjet, and atresic palate	N/A	NR	The prevalence of posterior crossbite was significantly increased in mouth breathers
Pacheco 2015 [14]	Class I occlusion: MB (74%) vs NB (79%) - ns Class II occlusion: MB (21%) vs NB (18%) - ns Class III occlusion: MB (5%) vs NB (4%) - ns The statistically significant findings (p < 0.050) were overjet greater than 4 mm and atresic palate	N/A	NR	In mouth breathers, the prevalence of an overjet >4mm and an atresic palate is significantly higher than in nasal breathers
Rossi 2015 [13]	Class I occlusion: MB (28%) vs NB (41%) - ns Class II occlusion: MB (64%) vs NB (48%) - p=0.001 Class III occlusion: MB (8%) vs NB (11%) - ns	N/A	Mouth breathing compared to nasal breathing: 2.02 (1.32 - 3.09)	There is an association between class II malocclusion and mouth breathing
Shanker 2004 [33]	% Overbite: T1: MB (44%) vs NB (42%) - ns T2: MB (34%) vs NB (41%) - ns T3: MB (41%) vs NB (53%) - ns T4: MB (42%) vs NB (52%) - ns Class I occlusion: MB (52%) vs NB (79%) - p<0.05 Class II occlusion: MB (44%) vs NB (18%) - p<0.05	N/A	NR	The respiratory mode had no statistically significant relationship to dentofacial morphology
Zicari 2009 [30]	Class III occlusion: MB (5%) vs NB (3%) - p<0.05 Crossbite: MB (34%) vs NB (15%) - p<0.05 Open bite: MB (15%) vs NB (25%) - p<0.001 Deep bite: MB (29%) vs NB (0%) - p<0.001	N/A	NR	The results showed a strong correlation between oral breathing and malocclusions

MB: Mouth breathers; N/A: Not applicable; NR: Not reported; NB: Nose breathers; ns: not significant; OR: Odds Ratio

4. Discussion

Results of the systematic review

The prevalence rates for class I to class III malocclusions vary significantly between studies with a prevalence between 9% and 74% for class I malocclusions, 21% and 73% for class II and between 5% and 24% for class III. These large percentage differences may be due to using different diagnostic criteria, as evident from the extracted data of the present review, as well as different methodologies used in the different studies. De Menezes et al (2006) [15] comments on different criteria for identifying mouth breathers, claiming it may be rare to find exclusively oral breathing patterns, hence the majority of patients most likely being of mixed breathing type [15]. This will make assessment very heterogeneous when no standardised guidance on the diagnosis of mouth

breathing is available. The guideline proposal for the clinical recognition of mouth breathing in children serves as an important step to standardise the assessment procedures so methods and study results can be compared in future research. [44]

Five out of the seven studies investigated the prevalence of class I – III malocclusion in mouth breathers compared to nasal breathers. The results are very heterogeneous and revealed that in mouth breathers, one out of five studies assessing the prevalence of class I – III malocclusion showed a significant difference for class I-III malocclusions compared to nasal breathers (Zicari 2009 [30]). Another study (Rossi 2015 [13]) showed a statistically significant difference between mouth and nasal breathers only in the prevalence of class II malocclusions, which also showed a moderate to poor quality rating. Two studies did not show a statistically significant difference between mouth and nasal

breathers for the occurrence of class I-III malocclusions (Harari et al., 2010 [31]; Pacheco et al., 2015 [14]). One further study showed trends of differences in malocclusion patterns between mouth and nasal breathers but did not conduct a statistical analysis (D'Ascanio 2010 [32]). Additionally, in children with malocclusions, only one study reported results on a significantly higher prevalence of mouth breathing compared to nasal breathing but did not differentiate between different classes of malocclusion.

These results are somewhat discrepant from the results of a previously conducted systematic review by Fraga et al. (2018) to assess the prevalence of dental malocclusion in mouth breathing children. [26] This review considered studies published up to 2014, reporting on prevalence data of malocclusion in mouth breathing children. The review authors identified seven articles, which collectively showed that the prevalence of malocclusion of Angle Class II, division 1 was higher than class I in mouth breathers. However, out of these seven studies, only two studies used control groups. [26] Additionally, the majority of studies in Fraga et al. (2018) were Brazilian studies, [26] which have not been included in the present review due to language restrictions. Since there seem to be a large number of Portuguese-language studies on the topic of mouth breathing and malocclusion available in the literature which have not been included in the present review, it may well be that the results may then only show half of the story. Future reviews should make any effort to include the major languages for article selection. Additionally, as Fraga et al. (2018) already highlighted, the research that came out in the last years on the topic is scarce. [26] This present review confirms that; no English language article was identified after the year 2015. The reasons for that can only be speculated upon. Fraga et al. (2018) explains this with the necessity for a multidisciplinary approach in the treatment of these children, with the presence of various healthcare professionals, such as orthodontists, otolaryngologist, speech therapist, allergist, physiotherapist and others, hence, impairing the ability to conduct research on the topic. [26]

To get a more realistic picture of the prevalence of malocclusion in mouth breathers in comparison to nose breathers, the present review defined as one of the key eligibility criteria the presence of a control group of nose breathers. Only with data from this control group can a comparison and a subsequent conclusion be made whether there is actually a difference in the occurrence of malocclusion. Currently, there is the assumption in the literature that mouth breathing might be associated in general with an increased occurrence of malocclusion or with an increase of the severity of the malocclusion. Additionally, the cause-and-effect relationship is unclear, with some authors claiming that mouth breathing might cause malocclusion and others stating the opposite. These assumptions are generally based on single studies that have been conducted more than a decade ago when research standards only just started to emerge, and reporting of the studies was most likely suboptimal. Only recently, Zhao et al. (2020) has conducted a

systematic review and meta-analysis to assess the effect of mouth breathing on facial skeletal development and malocclusion in children [16]. The study authors included all evidence published up to February 2020. A total of seven studies were included. The results of this review revealed that mouth breathing can be associated with an underdevelopment of the mandible, where it rotates backward and downward, and a steep occlusal plane [16]. Notably, there was little effect on the maxilla and a tendency of lip inclination in mouth breathing children. This, to date, is the first systematic review and meta-analysis that assessed this relationship [16]. It is an interesting finding which can draw conclusions on the severity of the malocclusion, but it cannot give an indication of whether the prevalence of malocclusion is indeed increased due to the mouth breathing.

The authors of this review concluded that mouth breathing can cause underdevelopment of the mandible [16]. It is unclear how this statement has been arrived at when the author of this review did not assess studies where the aetiology of mouth breathing has been removed, and subsequent assessments have taken place to compare it with a control group. Other studies have shown that facial skeletal development can be improved after the causes of the mouth breathing have been addressed by surgery or other methods [19, 34, 35]. Due to insufficient and low-quality evidence, the quest for the cause-and-effect relationship is still going on with further high-quality studies needed to draw adequate conclusions.

When considering the results of the present review where a clear conclusion with regards to increased prevalence of malocclusion in children with mouth breathing could not be drawn due to insufficient evidence, one could speculate that given most of the studies in the present review did not show a difference between nasal and mouth breathers, that indeed only the severity of malocclusion might be increased in mouth breathers, not the prevalence itself. This would tie in with some of the above-mentioned and other studies where surgery had an effect on arch width and malocclusion being partially reversed, indicating that environmental factors play at least a role in the development of malocclusion in mouth breathers [19, 34, 35, 37].

Studies on twins with malocclusion also confirm these observations of an environmental component in the development of malocclusion. For example, Jena et al. (2005) reports on a case of monozygotic female twins with similar dentition but the occlusions differed to some extent, where twin 1 had more severe reverse overjet, overbite and class III malocclusions compared to the other twin. [45] There were also marked differences in the cranio-dental-facial structures of the twins, such as the anterior-posterior position of the mandible, with twin 1 showing a more forward position than twin 2, the posterior facial height was higher in twin 2 compared to twin 1, flat cranial base of twin 2, and the length of maxilla and mandible was smaller in twin 2 compared to twin 1. [45] The position of the upper but not lower lip was also different between the twins. Due to the differences in the anterior-posterior position of the mandible,

Jena et al. (2005) conclude that this must have occurred due to environmental reasons. [45] Additionally, the different lengths of maxilla and mandible confirmed previous findings where other authors have also shown this phenomenon, indicating that these features play a significant role in the severity of class III malocclusion. [45] Further, the more severe overjet in twin 2 was interpreted as an environmental compensation in the variation of the severity of class III malocclusion. Hence, the authors concluded that environmental factors may play a significant role in the severity of class III malocclusion. [45]

Another case report on twins with class II malocclusion showed that these monozygotic brothers had similar facial appearance but their occlusions were remarkably different, with twin 1 showing a class II division 2 malocclusion, an overjet of 1 mm and a deep bite, and twin 2 displaying a class II division 1 malocclusion with a 12 mm overjet and a deep bite. [46] The mother reported finger sucking in twin 2 but not twin 1. According to the radiographic cephalometric analysis, there was no difference between the boys in the morphology but inclination of upper incisors differed, with the teeth retroclined in twin 2 compared to proclined teeth in twin 1. [46] In addition, twin 1 exhibited a high lip line. Both boys were treated successfully with removable and fixed orthodontics for four years, after which no difference in dentoskeletal morphology between the two twins was apparent. [46]

The included studies in the present systematic review were subject to high heterogeneity. This was reflected in different characteristics of the study participants, the methods of assessments, and the measuring instruments, as well as the inclusion and exclusion criteria of these studies. A key observation in the present work was that many of the information concerning the recruitment, eligibility, and demographic characteristics of the study participants were missing. It is, therefore, difficult to adequately estimate or analyse the results of these studies. Furthermore, due to the lack of the reporting of these data, these studies are unlikely to be repeatable in clinical practice.

Additionally, a wealth of instruments and tests were used to measure the prevalence of malocclusion, which is likely the result of a necessary multidisciplinary assessment before the diagnosis of malocclusion or mouth breathing can be formulated. None of the included studies mentioned the use of a quantitative assessment method for malocclusion. However, due to this heterogeneity, the analysis and comparability of the result were difficult. It was noted in the present review that the classification of malocclusion was not mentioned in all except one study. Fraga et al. (2018) in their review did not include studies other than those using the Angle classification. [26] A number of classification methods of malocclusion have been described in the literature, indicating that there seem to be no common standards or guidelines rather than these indexes and methods being based on opinions and preferences of single users. 1 Each one of these methods may have their advantages and disadvantages, with no particular method available that is inclusive of all

recommended criteria. [1] Two studies (Shanker 2004 and Zicari 2009) performed an intraoral measurement for the determination of malocclusion, without further details given. [30, 33] The use of intra-oral assessment as a reliable method for detection of malocclusion has been investigated by Ovsenik (2007), with the aim to investigate whether there were significant differences between intraoral measurements compared to measurements via study casts. [47] This study was performed on 92 children aged 14 years, in which the occlusal anomalies were detected intraorally as well as by taking study casts. The results of this study showed almost complete agreements between the two measurements for anterior crossbite, anterior open bite, transverse occlusion of the posterior teeth, and crowding. [47] Excellent reliability was shown for rotation of the incisors and canines, buccal segment relationship, overjet, and axial inclination of teeth. [47] The overall classification into malocclusion severity grades resulted in excellent agreement between the two methods, indicating that the intraoral measurement can reliably be used in screening and epidemiological studies. [47]

The authors of the present review has not identified a systematic literature review on malocclusion assessment methods and an analysis of their validity, reliability, and use within the research community. This would be a recommendation for future research in this area to get an overview of which methods have been used to date and how reliable and valid they are, which could give further indications on the research needs in this area and where to focus the research efforts.

For the assessment of mouth breathing, again a variety of methods were presented in the included studies. Only two studies used the mirror test, one study used two types of the lip seal test, and two studies asked questions to parents or the child. Only three studies reported on a physical assessment. For the remainder of the studies, this information was not available. Although not reported, it is likely that visual or physical assessment might have occurred. The in 2015 published guideline proposal for the clinical assessment of mouth breathing in children recommend at least two breathing tests with the child sitting and at rest. For future studies in this area, it is recommended to include more detail on the assessment methods, including what exactly was measured, how it was measured, when, and by whom it was measured. This would ensure better repeatability and judgement of results, as is currently possible due to insufficient reporting.

As part of the background research for this dissertation it was noted that prevalence rates for mouth breathing between studies differ significantly. Currently, the results of the present systematic review on the prevalence of malocclusion in mouth breathers do not give away an estimation on the total population of children affected, as no clear indication of prevalence rates of mouth breathers has been established. Since no systematic literature research has been identified on the topic of prevalence of mouth breathing, this would be a welcome future research project that could serve to get an

overview of the prevalence of mouth breathing and guide future developments on the topic.

In addition, large differences in sample sizes were identified between the studies. The largest study included more than 900 study participants (Rossi 2015) [13], while the Study by Harari et al. (2010) included only 116 study participants [31]. These discrepancies may have an impact on the comparability of the results, as drop out of subjects may impact on the final study results and the analysis.

Furthermore, the study participants comprised of children with large age gaps, with most studies assessing older children; only one study assessed children in the age range 3-6 years (Gois 2008). Future studies should make an effort to stratify results by age as the prevalence of malocclusion in younger mouth breathing children may be different compared to older children.

With regard to the generalisability of the study results, the current results may only be generalisable to high- to middle-income countries of unclear ethnicity and only in older children. Future research needs to make efforts to include and report on different ethnicities in research as well as ages and study countries in order to draw conclusions for the general paediatric population.

In order to assess the methodological quality of the studies, many lacked the necessary information, e.g. on the method of participant recruitment and inclusion and exclusion criteria of the study population. Additionally, the role of confounders generally has not been taken into account. Confounding factors in these studies could have been age, gender, ethnicity or socio-economic status. These have rarely been reported upon or taken into account in the statistical analysis plan. Future studies should address these shortcomings by paying attention to accurate reporting of study and methodological characteristics.

5. Conclusion and Future Recommendations

In conclusion, the results of this systematic literature review highlighted that research on the prevalence of malocclusions in mouth breathing children compared to nasal breathing children is scarce. Identified prevalence rates differ significantly, which is likely due to different assessment and reporting methods used in the included studies.

Additionally, in mouth breathing children, there is no clear evidence of an increased prevalence of malocclusion compared with nose breathers.

Future research would benefit from assessing the overall evidence on the prevalence of mouth breathing and malocclusion in children, as well as common assessment methods of malocclusion in a systematic review to provide recommendations for further clinical research in this area.

In addition, studies should improve the reporting of the research by adhering to the EQUATOR guidelines for observational studies in better describing the assessment and participant recruitment methods. Additionally, aspects of the

methodology should also be improved, for example, the consideration of confounding factors in the statistical analysis.

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